Review Article

Pigmented Villonodular Synovitis Presenting as Unilateral Hearing Loss: Review of the Literature and Case Report

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Keywords

Pigmented villonodular synovitis · Unilateral hearing loss · Temporomandibular joint

Abstract

Background/Aims: To review the existing literature on pigmented villonodular synovitis (PVNS) of the temporomandibular joint (TMJ) and report a rare case of PVNS of the TMJ presenting with unilateral hearing loss. **Methods:** Review of the existing literature and a description of personal experience with PVNS of the TMJ presenting with unilateral hearing loss. **Results:** Review of the existing literature revealed 76 reported cases of PVNS of the TMJ. The most common presenting symptom was of a slowly enlarging mass or swelling of the preauricular area, with dysfunctional TMJ also frequently reported. All patients underwent surgical excision with some pursuing radiation as adjuvant therapy. **Presented Patient:** A 46-year-old man presented with several months of unilateral subjective hearing loss and aural fullness. Imaging revealed a mass centered along the superior TMJ with expansion through the squamous temporal bone and extra-axial intracranial extension into the middle cranial fossa. Imaging characteristics and fine-needle aspiration biopsy were consistent with PVNS. **Intervention:** The patient underwent near-total excision of the mass via frontotemporal craniectomy and lateral temporal bone resection. **Follow-Up:** At the 16-month follow-up there was no evidence of disease recurrence. **Conclusion:** PVNS of the TMJ represents a rare entity that can present with a variety of symptoms including unilateral hearing loss.

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Introduction

Pigmented villonodular synovitis (PVNS) is an intra-articular, proliferative process of the synovium also referred to as a diffuse-type giant cell tumor by the World Health Organization (WHO) [1]. It is rare, with an overall incidence of 1.8 cases per million in the USA. Generally a monoarticular process of the large joints, PVNS affects the knee in about 80% of cases, with the hip, ankle, shoulder, and elbow the next most common sites [1, 2]. Men and women are equally affected, usually occurring between the age of 20 and 50 years [3]. Although the underlying pathogenesis has been much debated, recent findings have favored a neoplastic etiology [4].

We present a thorough review of the existing literature as well as a rare case of a PVNS lesion of the temporomandibular joint (TMJ) with middle cranial fossa and temporal bone invasion presenting with the primary complaint of hearing loss and without symptoms of TMJ dysfunction.

Results

Review of the Literature

The TMJ is rarely involved, with only 76 reported cases (Table 1). These cases represent about 3% of tumors or pseudo-tumors of the TMJ [5]. Even fewer involve invasion into the middle cranial fossa, with only 27 reported cases (Table 2). The most common presenting symptom is of a slowly enlarging mass or swelling of the preauricular area, with sequelae of a dysfunctional TMJ also frequently reported [6]. Erosion into the middle fossa was reported in 33 cases, however only 9 reported involvement of the dura. Hearing loss was reported in 14 cases. There were 10 cases of reported recurrence after surgical excision, with 2 of these having multiple recurrences. All patients underwent excision of their tumors. Six patients underwent radiation either as adjuvant therapy for subtotal resection or for recurrence of disease [7–12]. Two cases of malignant spread to the lung have been described, one resulting in death [8, 13].

Case Report

In October of 2013, a 46-year-old male presented with 3 months of aural fullness and unilateral subjective hearing loss. He had no history of trauma to the face or jaw, and no trismus. After several unsuccessful rounds of treatment for otitis media, he was referred to an otolaryngologist who noted complete occlusion of his ear canal by a smooth, epithelial-covered lesion. A CT of the temporal bone was performed which revealed an invasive skull base mass. He was referred to our center for further work-up and treatment. Physical examination revealed only a firm soft-tissue mass occluding the right external auditory canal. Audiologic testing revealed a moderately severe rising to mild sloping to profound mixed (mostly conductive) hearing loss. Monosyllabic word recognition was 92% when stimuli were presented at a loud listening level. Hearing in the left ear was essentially normal (Fig. 1). MRI with gadolinium and a noncontrast CT revealed a lobular, demarcated 5-cm mass centered along the superior TMJ with expansion into the temporalis muscle and extra-axial intracranial extension into the middle cranial fossa (Fig. 2). An ultrasound-guided fine-needle aspiration biopsy was performed revealing numerous multinucleated giant cells, hemosiderin-laden macrophages, and lymphocytes in a background of granulation tissue (Fig. 3). A presumed diagnosis of PVNS of the TMJ was made based on cytopathologic and radiologic findings.

Treatment

In February of 2013, the patient underwent surgical resection. Near-total removal of the tumor was performed through an extended postauricular incision that was carried superiorly

Table 1. All reported cases of PVNS of the TMJ

Patient	Year [Ref.]	Age/sex	Middle fossa	Dura	Temporal bone	Hearing loss	EAC
1	1973 [33]	22/M	N	N	N	NR	NR
2		58/F	N	N	N	Y	Y
2	1977 [34]	58/F	Y	Y	Y	Y	Y
3	1975 [35]	37/M	N	N	Y	NR	N
4	1977 [36]	34/M	N	N	N	NR	NR
5	1977 [37]	62/F	N	N	N	N	N
6	1978 [38]	55/F	N	N	N	NR	N
7	1981 [39]	36/M	N	N	N	NR	NR
8	1982 [40]	39/F	N	N	N	N	NR
9	1982 [41]	47/F	N	N	N	N	N
9	1983 [42]	47/F	N	N	Y	NR	NR
10	1984 [7]	61/F	N*	N	N*	NR	N*
10	1997 [8]	71/F	N*	N*	N*	NR	NR
11	1989 [43]	32/M	N	N	N	N	NR
12	1992 [44]	50/F	Y	Y	Y	Y	Y
13	1993 [45]	10/F	N	N	N	NR	NR
14	1994 [46]	59/F	N	N	N	NR	NR
15	1996 [47]	36/M	Y	Y	Y	Y	N
16	1996 [48]	41/F	N	N	Y	NR	Y
17	1997 [49]	39/F	NR	NR	NR	NR	Y
18	1997 [25]	47/M	N	N	N	NR	N
19	1997 [50]	70/F	N	N	N	NR	N
20	1997 [51]	48/M	N	N	N	NR	N
21	1998 [52]	42/F	N	N	Y	NR	N
22	1998 [53]	18/M	N	N	N	NR	NR
23	1999 [31]	37/M	Y	Y	Y	Y	Y
23	2001 [32]	35/M	Y	NR	Y	Y	Y
23	2005 [28]	36/M	Y	NR	Y	Y	Y
24	1999 [54]	57/F	N	N	N	NR	N
25	1999 [55]	63/M	Y	NR	Y	NR	NR
26	2000 [30]	59/F	N	N	N	NR	NR
27	2001 [56]	45/F	N	N	N	NR	N
28	2002 [57]	36/M	Y	Y	Y	Y	Y
29	2003 [58]	42/M	Y	NR	Y	Y	Y
30		33/M	N	N	N	NR	NR
31	2003 [59]	45/M	N	N	N	NR	NR
32	2004 [60]	33/M	Y	NR	Y	NR	NR
33	2004 [24]	28/F	Y	NR	Y	NR	NR
34		22/M	Y	NR	Y	NR	NR
35		47/M	N	N	N	NR	NR
36		58/M	N	N	N	NR	NR
37	2004 [61]	60/M	Y	NR	Y	Y	Y
38	2005 [62]	38/F	N	N	N	NR	NR
39	2005 [28]	36/F	N	N	N	NR	NR
40	2007 [27]	52/M	NR	NR	Y	NR	Y
41		67/M	Y	NR	Y	NR	NR
42	2008 [9]	56/F	Y	N	Y	Y	NR
43	2008 [63]	78/M	N	N	N	NR	NR
44	2008 [64]	38/M	Y	N	Y	NR	NR
45	2009 [29]	21/F	N	N	N	NR	NR
46	2009 [65]	36/M	Y	N	N	NR	NR
47	2010 [66]	22/F	N	N	N	NR	NR
48	2011 [67]	74/F	N	N	N	NR	NR
49		62/F	N	N	N	NR	NR
50		21/F	N	N	N	NR	NR

Table 1 (continued)

Patient	Year [Ref.]	Age/sex	Middle fossa	Dura	Temporal bone	Hearing loss	EAC
51		25/M	N	N	N	NR	NR
52	2011 [17]	59/F	Y	N	Y	Y	Y
53	2011 [26]	42/M	N	N	N	NR	NR
54	2011 [13]	26/M	Y	Y	Y	Y	NR
55	2011 [16]	29/M	Y	Y	Y	N	NR
56	2012 [68]	80/M	N	N	N	N	Y
57	2012 [67]	NR/NR	NR	NR	NR	NR	NR
58	2012 [14]	56/F	Y	N	Y	Y	NR
59	2012 [69]	51/F	N	N	N	NR	NR
60	2013 [6]	48/M	N	N	N	NR	NR
61	2014 [16]	53/F	N	NR	NR	NR	NR
62		38/F	Y	NR	NR	NR	NR
63		31/M	Y	NR	NR	NR	NR
64		49/M	N	NR	NR	NR	NR
65		50/F	Y	NR	NR	NR	NR
66		34/M	Y	NR	NR	NR	NR
67		48/M	Y	NR	NR	NR	NR
68		56/F	Y	NR	NR	NR	NR
69	2014 [70]	38/M	N	N	N	NR	N
70	2014 [12]	55/M	Y	Y	Y	NR	N
71		26/F	Y	NR	Y	NR	N
72		58/M	Y	NR	Y	NR	N
73		52/M	Y	NR	Y	NR	Y
74		35/M	N	N	Y	NR	N
75	2015 [71]	47/F	Y	N	Y	NR	N
76	2015 [72]	51/M	Y	Y	Y	NR	N

EAC, external auditory canal; Y, involved; N, not involved; NR, not reported; N*, not involved on initial presentation, but present on recurrence.

Table 2. Incidence of bony involvement in reported PVNS cases

Middle fossa involved	33 (43)
Temporal bone involved	32 (42)
Dura involved	9 (12)
EAC or TM findings	15 (20)
Hearing loss	14 (18)

Values are *n* (%). EAC, external auditory canal; TM, tympanic membrane.

to allow for a frontotemporal craniotomy, and inferiorly to allow for control of the great vessels in the neck. A lateral temporal bone resection was performed as a gross tumor was noted in the middle ear with erosion of the incus. The tumor was found to extend into the eustachian tube with dehiscence of the carotid artery.

The capsule of the TMJ was incised, and inspection of the joint space revealed a localized tumor in the posterosuperior aspect, but there was no evidence of diffuse joint disease. After removal of the tumor, no further excision of the joint was required.

The involved temporalis muscle overlying the squamous temporal bone was removed. A frontotemporal craniectomy was performed with debulking of the intracranial portion

Fig. 1. Preoperative audiogram showing a moderately severe rising to mild sloping to profound mixed (mostly conductive) hearing loss in the right ear with normal hearing in the left ear.

Fig. 2. Pap-stained smear (200×) showing numerous multinucleated giant cells with surrounding mononuclear histiocytes/macrophages. Some of the macrophages are pigmented (arrows).

of the tumor where it was found to densely adhere to, but not invade, the dura. The majority of the tumor was removed; however, residual disease was left at the most adherent portions of the dura and along the course of the petrous portion of the carotid artery.

The defect was repaired with an anterolateral thigh free flap. During dissection of the neck for exposure of vessels, several enlarged, dark lymph nodes were excised. A limited suprahyoid neck dissection was performed, and frozen section analysis of one node found only reactive lymphoid tissue.

Fig. 3. Axial (a) and coronal (b) T1-weighted MRI showing a lobular demarcated mass centered in the right squamosal temporal bone along the top of the TMJ (dashed arrow) with expansion into the temporalis muscle (thin arrow), and with intracranial extra-axial extension into the middle cranial fossa (thick arrow).

Fig. 4. Low-power H&E stain of the resected mass showing numerous multinucleated giant cells, hemosiderin-laden macrophages, and lymphocytes present in a background of granulation tissue.

Follow-Up

The patient recovered from the surgery without neurologic deficits and was discharged from the hospital on postoperative day 7. Final pathology revealed a diffuse, extra-articular tenosynovial giant cell tumor (Fig. 4). One lymph node was positive for hemosiderin-laden macrophages.

Follow-up at 16 months revealed no evidence of recurrence. A small focus of enhancing tissue medial to the TMJ noted on MRI may represent residual tumor, but has thus far remained stable. The patient is asymptomatic except for occasional headaches and the development of first bite syndrome. First bite syndrome is a short-lived severe spasming over the parotid region with the initiation of chewing which is often a complication of injury to the sympa-

thetic innervation of the parotid from infratemporal fossa surgery. This was successfully managed with Botox injections. Radiation therapy is being reserved for evidence of progression.

Discussion

First described as a nodular lesion of the synovial membrane in 1852, it was not until 1941 that the term PVNS was applied and the current description of the tumor was published [14, 15]. The WHO now uses the term diffuse-type giant cell tumor for the diffuse intra-articular form of PVNS; however, many clinical reports continue to use the original terminology [16].

The underlying pathogenesis of PVNS has been difficult to elucidate. Since the original report by Jaffe et al. [15], PVNS has been thought to represent an inflammatory response to an unknown insult. Experiments using animal models were able to recreate PVNS-like lesions through repeated infusion of blood or iron solutions, suggesting trauma as a contributing factor; however, a review of reported cases in the TMJ found no consistent reports of preceding trauma [17–20]. The inflammatory hypothesis was supported by studies showing polyclonal cellular populations [21, 22]. In 2006, West et al. [4] showed that there is a small, but distinct monoclonal population of cells with a specific translocation in PVNS. They hypothesized that this neoplastic process was the underlying cause of the lesion and that it created a “tumor-landscaping” effect. This recruited an abundance of polyclonal inflammatory cells creating the typical histologic appearance and explaining the previous findings of a polyclonal population. Although not definitive, these findings, along with several others, strongly indicate an underlying neoplastic process stemming from a possible inflammatory insult, leading to the clinical and histologic findings of PVNS [1, 3].

Imaging can be effectively diagnostic for PVNS [23]. Lesions generally present on CT as an enhancing hyperdense soft tissue mass. Erosion is common in joints where the bony confines do not allow for uninterrupted expansion of the mass [1]. In PVNS originating in the TMJ, there is often bony destruction of the mandibular condyles. Skull base erosion with intracranial extension is less common [16]. MRI has become an important tool in the diagnosis and follow-up of this disease, and is the preferred imaging modality for the evaluation of PVNS [24]. PVNS has a distinct appearance on MR including low signal intensity on T2 images and a magnetic susceptibility artifact (blooming) on gradient-echo images due to the high hemosiderin concentration of the lesion [23]. Similarly, they often have low to intermediate signal intensity relative to muscle on T1, but can appear bright depending on their lipid content [1, 25]. Enhancement with gadolinium is common, but variable [23].

Bony destruction extending from the TMJ into the middle ear space can lead to hearing loss as seen in this case. Of the 76 cases in the literature, 20 described the patient’s hearing status. Fourteen (70%) of those cases had some degree of hearing loss. Only 4 cases described the severity and type of hearing loss that was present. Intriguingly, all the cases, mirroring our patient, had a severe conductive hearing loss. It is unknown if the remaining 56 cases had subclinical hearing loss and thereby did not prompt an audiogram to be performed.

Pathologic evaluation often reveals a diffusely thickened and inflamed synovium. When the lesion is localized to the joint space, the histology has a villous pattern; however, it becomes more multinodular with extension into surrounding tissues [26]. Rarely, chondroid metaplasia has been described in PVNS [27]. The brownish coloring of the tumor often noted at surgery is due to hemosiderin, which can be variable. Lesions are composed of mononuclear histiocytes, multinucleated giant cells, and xanthoma cells in varying proportions [23]. Fine-needle aspiration can effectively diagnose PVNS. Findings include single and loose

clusters of mononuclear and multinucleated giant cells. Mild to moderate atypia may be present but is not beyond that expected for a reactive process [26].

Total surgical resection is the treatment of choice for PVNS [6]. Some have advocated total synovectomy to prevent recurrence; however, many PVNS tumors have been removed with more conservative measures (e.g., arthroscopy) with favorable long-term follow-up [28, 29]. It is for this reason that it was decided to maintain the TMJ as conservative management has been shown to be able to control the disease, and there was limited disease within the joint. Additionally, the options for TMJ reconstruction are all sub-par, and therefore the team planned close surveillance. Tumors with intracranial extension have undergone extensive surgical exploration into the middle fossa and infratemporal fossa similar to the case presented here. Even with these extensive resections, there are only 2 cases of facial nerve sacrifice [28, 30].

As this lesion is not a malignant neoplastic process with metastatic potential, there is no need for a neck dissection to look for occult disease. The reason for a neck dissection in this case was twofold. First, the extensive defect required a free flap reconstruction necessitating the exposure of neck vessels. Secondly, the patient had lymphadenopathy and due to the rarity of PVNS there was some question about the validity of the fine-needle aspiration diagnosis.

Although generally highly vascular, there is only one report of preoperative embolization being utilized [28, 31, 32]. Radiation has been utilized for subtotal resection, recurrence of disease, or especially infiltrative tumors [8–11]. Treatment outcome data are difficult to interpret due to the low number of cases, and treatment decisions often must be made based on inferences from PVNS at other sites. As such, developing an evidence-based surveillance protocol is difficult. Nonetheless, we are repeating MRI every 6 months for the first 2 years and then spacing out imaging to yearly or sooner if the patient becomes symptomatic. Larger case series in other joints have shown that there is a first-year recurrence rate as high as 32% and recurrences can occur as long as 16 years later [73].

Conclusion

The presented case represents an unusual presentation of a rare disease. The patient did not have the typical preauricular swelling and showed no evidence of TMJ dysfunction. His primary presenting symptom was hearing loss due to the tumor's direct involvement of the external auditory canal and middle ear. Thus, taken with the review of the literature, PVNS of the TMJ represents a rare entity that can present with a variety of symptoms including unilateral hearing loss.

Statement of Ethics

This research was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. The patient consented to having her case report published.

Disclosure Statement

The authors have no funding, financial relationships, or conflicts of interest to disclose.

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